Condition Severity and Psychosocial Functioning in Pre-Adolescents With Spina Bifida: Disentangling Proximal Functional Status and Distal Adjustment Outcomes

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Objective: To examine relations between condition severity and psychosocial functioning in 70 8- and 9-year-old pre-adolescents with spina bifida by testing several direct, indirect, and mediated effects models for proximal functional status and distal adjustment outcomes.

Methods: Proximal functional status outcomes (e.g., degree of involvement in activities, scholastic competence, athletic competence, attentional problems) and distal adjustment outcomes (e.g., behavior problems, social competence) were assessed with mother, father, and teacher report. Severity variables included spinal lesion level, spina bifida classification, shunt status, ambulation status, number of shunt surgeries, and two severity composites.

Results: Condition severity was associated with the proximal functional status outcomes across parent and teacher report. In contrast, no significant relationships were found between the severity parameters and distal adjustment outcomes. Findings supported a proximal effects model of condition severity as well as an indirect effects model (e.g., presence of a shunt → less scholastic competence → less social competence) and were consistent with recent theoretical formulations (e.g., Wallander & Varni, 1995).

Conclusions: Disentangling proximal functional status outcomes and distal adjustment outcomes is critical in studies of condition severity and psychosocial functioning. We discuss clinical implications.

Key words: spina bifida; physical disability; condition severity; child adjustment; functional status; proximal; distal.

Numerous studies indicate that children with physical disabilities and chronic illnesses are at increased risk for experiencing psychosocial adjustment problems (Appleton et al., 1997; Lavigne & Faier-Routman, 1992; Thompson & Gustafson, 1996; Wallander & Thompson, 1995). Moreover, investigators have examined factors that may be associated with adjustment in these populations, including child temperament, child and parent coping strategies, cognitive processes, stressors, family functioning, and social support (Lavigne & Faier-Routman, 1993; Thompson & Gustafson, 1996; Wallander & Thompson, 1995). Disease (or condition) parameters have also been investigated in relation to child adjustment in various populations.
Associations between condition severity and psychosocial adjustment in children with spina bifida are often weak and nonsignificant (Wallander, Feldman, & Varni, 1989; Wallander, Varni, et al., 1989; although see Holmbeck & Faier-Routman, 1995), leading some to conclude that variation in physical status does not account for a significant portion of the observed variation in adjustment (Wallander, Feldman, et al., 1989). However, studies examining associations between severity and adjustment in other pediatric populations have yielded mixed findings (Lavigne & Faier-Routman, 1993; Thompson & Gustafson, 1996). Based on a theoretical model of maladjustment in children with chronic illnesses and physical disabilities, Wallander and Varni (1995) suggest that condition severity may be directly associated with child adjustment (i.e., mental health and social functioning), but may also be related to adjustment indirectly via its impact on the functional status of the child. Functional status has been defined as “the degree to which the child [can] perform daily tasks at an age appropriate level” (Lavigne & Faier-Routman, 1993, p. 119). “[G]ood functional status is demonstrated by a child who exhibits the full range of age-appropriate physical, cognitive, emotional, and social behaviors” (Dadds, Stein, & Silver, 1995, p. 529).

The lack of significant severity → adjustment associations in most studies may be due to researchers’ failure to (1) examine the differential impact of multiple severity parameters, and (2) differentiate between (and include measures of) proximal functional status outcomes and distal adjustment outcomes. For the first point, even when multiple illness parameters are examined, investigators tend to use composite severity scores rather than examining the parameters individually, even though findings suggest that different condition parameters may be associated with different outcomes (e.g., Holmbeck & Faier-Routman, 1995). For the second point, choice of outcome variables in our study was based on the distinction between “proximal functional status outcomes” and “distal adjustment outcomes.”

As defined in this study, the proximal functional status outcomes represent functional consequences of specific disability-related symptoms and are, therefore, likely to vary as a function of condition severity. Given the nature of the orthopedic and neurological problems typically associated with spina bifida, the proximal functional status variables included an array of physical/athletic (i.e., activity level, athletic competence, perceived physical appearance, and somatic complaints) and cognitive outcomes (i.e., attention problems and scholastic competence; see Figure 1). Based on Wallander and Varni’s (1995) model, the distal adjustment variables included frequently studied mental health (i.e., internalizing and externalizing symptoms) and social functioning outcomes (e.g., Appleton et al., 1997; Varni & Wallander, 1988; Wallander, Feldman, et al., 1989; Wallander, Varni, et al., 1989; see Figure 1). These distal outcomes can be distinguished from the proximal outcomes insofar as the former (unlike the latter) do not clearly represent functional limitations related to spina bifida symptomatology.

Figure 2 presents seven models that illustrate possible associations between condition severity and the proximal and distal outcomes, including some that have been discussed by other investigators (e.g., models C and D are based on Wallander & Varni’s 1995 model). Simple effects (models A and B in Figure 1), indirect effects (models C and E), mediated effects (models D and F), and moderated effects models (model G) are differentiated in Figure 2 (see Baron & Kenny, 1986; Holmbeck, 1997, for more complete discussions of the terms mediation and moderation). Indirect effects models are path models whereby the predictor (e.g., shunt status) affects an intermediate variable (e.g., attention problems), which, in turn, affects a criterion variable (e.g., social problems). Mediated effects models are similar to indirect effects models except that the predictor has a direct effect on the criterion and some (or all) of this direct effect can be attributed to the mediational pathway. Finally, the moderator model suggests that severity is related to various outcomes but only under certain conditions, as specified by the level of the moderator variable (e.g., gender of child).

In our investigation, we tested the utility of models A, B, C, and D. To determine whether findings were consistent across family and school contexts, the models were tested twice, once with parent report of outcome and once with teacher report. We anticipated that the severity parameters (e.g., shunt status, lesion level, ambulation status, and type of spina bifida) would be associated with
the proximal functional status outcomes (model A in Figure 2), with the expectation that shunt status would be associated with the cognitive variables (e.g., Holmbeck & Faier-Routman, 1995), and lesion level and ambulation status would be associated with the physical/athletic variables. We expected condition severity to be related to the distal variables only indirectly via the proximal functional status outcomes (i.e., support for model C, but not model B, was expected; see Figure 2). Given our expectation that there would be no direct effect between the severity parameters and the distal outcomes, we did not expect model D to be supported. An evaluation of models C and D represents a partial test of Wallander and Varni's (1995) model of adjustment in pediatric populations.

**Method**

**Sample**

Participants were 70 families with 8- and 9-year-old pre-adolescents with spina bifida (39 males, 31 females; M [age] = 8.33 years; 80% white) who were participants in a larger longitudinal study of the transition to adolescence (Holmbeck et al., 1997).
The sample included a wide range of family annual incomes (33% < $40,000; 24% = $40,000–$49,999; 39% > $50,000; 4% missing) and maternal (M = 37.71) and paternal (M = 40.91) ages. Biological mothers from all families and 56 fathers participated in the study (79% two-parent, intact families; 7% mother/stepfather; 12% single-mother; 2% other). The mean Peabody Picture Vocabulary Test score (PPVT-R; Dunn & Dunn, 1981) was 91.70 (SD = 18.80). While the current sample differed from samples studied in previous investigations due to its narrow age range, the distributions of participants falling into other demographic categories or into the condition severity categories (see below) were comparable to distributions of other studies (i.e., Holmbeck & Faier-Routman, 1995; Wallander, Feldman et al., 1989).

Families participating in the study were recruited from lists provided by the following: (1) a children’s hospital, (2) a hospital that cares exclusively for children with physical disabilities, (3) a statewide spina bifida association, and (4) a university-based medical center. A comparison of participating children from families that declined to participate (n = 64) revealed no differences with respect to lesion level (χ² (2) = .62, p > .05) or type of spina bifida (myelomeningocele vs. lipomeningocele) (χ² (1) = 1.63, p > .05). The relatively high decline rate was attributed to the extensive time commitment asked of the participants (e.g., a multiwave longitudinal study consisting of several 3-hour home visits).

Procedure

Family assessments were conducted by trained research assistants during 3-hour home sessions. Parents and children were asked to complete a set of questionnaires and 1 hour of family interaction tasks that were videotaped and audiotaped, for which the family was paid $50.00. Child questionnaires were administered in an interview format with all Likert scales printed on large laminated cards. After the family visit, questionnaire packets were sent to the child’s teacher and a health professional, for which they were compensated with $5.00 and $2.00, respectively (teacher return rate = 97%; n = 68; health professional return rate = 98%, n = 69). This study involves analyses of mother, father, teacher, and health professional questionnaire data along with information obtained from medical chart reviews.

Measures

Internalizing and Externalizing Behavior Problems and Competence. Mother, father, and teacher reports on the Child Behavior Checklist (CBCL; Achenbach, 1991a) were used to evaluate behavioral adjustment and competence. The parent version of this measure is comprised of three competence scales, eight problem scales, and two second-order problem scales representing internalizing and externalizing behavior problems. The eight problem scales include withdrawn, somatic complaints, anxious/depressed, social problems, thought problems, attention problems, delinquent behavior, and aggressive behavior. Competence scales based on parental responses include activities, social competence, and school competence. In this study, seven of the eight problem scales and two of the three competence scales were examined. The thought problems subscale was not used because it did not fit adequately into any of the proximal or distal outcome categories in Table I (given the item content), and the school competence items were not included in the questionnaire because they were redundant with other items in the larger research protocol. The teacher report version of this measure, the Teacher Report Form (TRF; Achenbach, 1991b), was utilized to gather teacher-report information on the seven problem scales described above (the TRF does not include activities and social competence scales).

Perceived Self-Concept. Perceived child self-concept was assessed with mother, father, and teacher report on Harter’s (1985) Rating Scale of Child’s Actual Behavior. The 15-item Teacher’s Rating Scale of Child’s Actual Behavior (Harter, 1985) is based on the 36-item child-report version and was adapted for parent-report. The measure contains five three-item scales: scholastic competence, social acceptance, athletic competence, perceived physical appearance, and behavioral conduct. Alphas ranged from .75 to .84 for mother report, .69 to .81 for father report, and .82 to .93 for teacher report. All scores were converted to item-mean form.

Condition Parameters. Information on condition severity was obtained from reviews of medical charts, maternal reports, and health professional ratings. The following parameters were obtained from the medical chart: (1) lesion level (based on the median of the three most recent reports; 30% sacral, 54% lumbar, 12% thoracic, 4% missing; interrater reliability = 83%), (2) type of spina bifida (83% myelomeningocele; 11% lipomeningocele, 6% other;
Table 1. Analyses Examining Associations between Condition Severity and the Proximal Status Outcomes

<table>
<thead>
<tr>
<th>Condition Severity Variable</th>
<th>Cognitive</th>
<th>Physical/Athletic</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Parent</td>
<td>Teacher</td>
</tr>
<tr>
<td>Lesion level</td>
<td>$f(4, 118) = 2.81^*$</td>
<td>$f(4, 118) = .99$</td>
</tr>
<tr>
<td>Spina bifida classification</td>
<td>$f(2, 60) = 2.46$</td>
<td>$f(2, 60) = 1.85$</td>
</tr>
<tr>
<td>Shunt status</td>
<td>$f(2, 64) = 3.37^*$</td>
<td>$f(2, 64) = 3.03^*$</td>
</tr>
<tr>
<td>Ambulation status</td>
<td>$f(4, 124) = 1.21$</td>
<td>$f(4, 124) = 2.40^*$</td>
</tr>
</tbody>
</table>

Significant univariate results are noted below all significant MANOVAs. Schol comp = scholastic competence, ath comp = athletic competence, att probs = attention problems, ns = nonsignificant, U = unshunted; S = shunted, S = sacral, T = thoracic, L = lipomeningocele, and M = myelomeningocele. For ambulation status, 1 = no assistance; AFOs, 2 = KAFOs/HKAFOs, and 3 = wheelchair. Proximal parent outcomes are based on composites of mother and father data.

*p < .05.

**p < .01.

interrater reliability = 92%), and (3) total number of shunt surgeries (other than the original shunt placement surgery; $M = 2.50$; $SD = 2.91$; interrater reliability = .97). The following parameters were based on maternal report: (1) shunt status (71% were shunted), and (2) ambulation (43% required no assistance or ankle-foot orthoses [AFOs] to ambulate more than 50% of the time, 40% ambulated with knee-ankle-foot orthoses [KAFOs] or hip-knee-ankle-foot orthoses [HKAFOs] more than 50% of the time, and 17% ambulated with wheelchair assistance more than 50% of the time). None of the severity variables was associated in any systematic way with any of the demographic variables.

An overall severity composite score based on the four categorical mother-reported and medical chart severity variables was computed for each participant, with scores ranging from 4 to 10 (higher scores index higher levels of severity). These scores were based on the participant’s inclusion in a specific group for the following variables (α = .70): shunt status (no = 1, yes = 2), myelomeningocele (no = 1, yes = 2), lesion level (sacral = 1, lumbar = 2, thoracic = 3), and ambulation status (no assistance/AFOs = 1, KAFOs/HKAFOs = 2, wheelchair = 3).

Finally, health professional ratings of severity were assessed across the following six functioning areas on a Likert scale from 1 (low severity) to 5 (high severity) for each child: orthopedic, neurological, urinary, bowel, behavior and mobility, and overall condition (α = .91). In support of the validity of the severity variables, the severity composite based on maternal and medical chart data was significantly associated with the health professional severity composite ($r = .60$, $p < .001$).

**Data Reduction**

In order to reduce the number of potential analyses (given the number of respondents and variables), mother- and father-reported outcomes were combined for all families where father data were available (otherwise mother data were used). Such data reduction was justified based on mother-father intercorrelations: .31 to .61 ($M = .47$) for the CBCL problem scales, .49 to .54 ($M = .52$) for the two CBCL competence scales, and .20 to .70 ($M = .45$) for the Harter subscales. Although several of the condition severity predictors were significantly intercorrelated, they were analyzed separately in this study, given past results suggesting that findings differ across condition parameters (Holmbeck & Faier-Rutman, 1995; Wallander, Feldman, et al., 1989) and because there are clear differences across the parameters in whether they are observable to other individuals and whether they are associated with neurological complications. On the other hand, because of the high intercorrelations and because past research has included composite severity scores
(e.g., Wallander, Feldman, et al., 1989), we employed two composite severity variables for comparative purposes.

**Analysis Plan**

To determine the utility of the simple proximal and distal models (models A and B, respectively, in Figure 2), four MANOVAs (based on the two proximal outcome variable sets and the two distal outcome variable sets; see Figure 1) were run for each of the four categorical independent variables (i.e., lesion level, spina bifida classification, ambulation status, and shunt status). These analyses were repeated for parent and teacher report. Pearson \( r \) correlations were computed to examine relations between the continuous predictors of severity (i.e., number of shunt surgeries, two severity composites) and the proximal and distal outcomes. The utility of the indirect and mediated effects models (models C and D in Figure 1) were evaluated with path analytic techniques (Cohen & Cohen, 1983) via multiple regression analyses.

**Results**

**Model A: Severity \( \rightarrow \) Proximal Functional Status Outcomes**

**Lesion Level.** Consistent with the proximal functional status hypothesis, the MANOVA that examined associations between lesion level and parent-report of the physical/athletic outcomes was significant, \( F(8, 114) = 2.34, p < .05 \) (see Table I). Follow-up univariate tests (see Table I) indicated that children in the sacral group (\( M = 43.22 \)) participated in more activities than children in the thoracic group (\( M = 37.06 \), \( F(2, 61) = 3.17, p < .05 \)). The MANOVA for associations between lesion level and parent-report of the cognitive outcomes was also significant, \( F(4, 118) = 2.81, p < .05 \), but none of the univariate follow-up effects were significant. The two MANOVAs for teacher-reported cognitive and physical/athletic outcomes were not significant.

**Spina Bifida Classification.** The two MANOVAs that examined the relationship between spina bifida classification and parent- and teacher-reported physical/athletic outcomes were significant: \( F(4, 58) = 3.72, p < .01 \); and \( F(3, 55) = 4.78, p < .01 \), respectively. Univariate follow-up analyses (see Table I) indicated that children with lipomeningocele (\( M = 47.44 \)) were more likely to participate in activities (according to parents) than children with myelomeningocele (\( M = 40.52 \)), \( F(1, 61) = 9.16, p < .01 \). Similarly, children with lipomeningocele (\( M = 2.62 \) and 2.69 for parent and teacher report, respectively) demonstrated higher levels of athletic competence than children with myelomeningocele (\( M = 2.02 \) and 1.73, respectively): parent-report, \( F(1, 61) = 8.59, p < .01 \); teacher-report, \( F(1, 57) = 13.23, p < .001 \).

**Shunt Status.** For shunt status, the two MANOVAs for the parent- and teacher-reported cognitive outcomes were significant: \( F(2, 64) = 3.37, p < .05 \); and \( F(2, 64) = 3.03, p < .05 \), respectively. Follow-up analyses (see Table I) indicated that children with shunts (\( M = 2.64 \) and 2.23 for parent and teacher report, respectively) demonstrated less scholastic competence than children without shunts (\( M = 3.10 \) and 2.68, respectively): parent-report, \( F(1, 65) = 6.85, p < .01 \); teacher-report, \( F(1, 65) = 5.44, p < .05 \). Children with shunts (\( M = 59.49 \)) also demonstrated more teacher-reported attention problems than children without shunts (\( M = 56.00 \)), \( F(1, 65) = 4.46, p < .05 \). Finally, the MANOVA for teacher-reported physical/athletic outcomes was significant, \( F(3, 59) = 2.84, p < .05 \). Follow-up analyses revealed that children with shunts (\( M = 1.74 \)) exhibited lower levels of athletic competence than those without shunts (\( M = 2.26 \)), \( F(1, 61) = 6.55, p < .01 \).

**Ambulation Status.** For ambulation status, the MANOVA for the parent-reported physical/athletic outcomes was significant, \( F(8, 120) = 2.06, p < .05 \). Follow-up analyses (see Table I) revealed that children using no assistance/AFOs or KAFOS/HKAFOs (\( M = 42.75 \) and 42.06, respectively) participated in more activities than children ambulating with a wheelchair (\( M = 36.18 \)), \( F(2, 64) = 5.21, p < .01 \). The MANOVA for the teacher-reported cognitive outcomes was also significant, \( F(4, 124) = 2.40, p < .05 \). Univariate analyses revealed that teacher-reported scholastic competence was higher among children who used no assistance or AFOs (\( M = 2.66 \)) than among children who used KAFOS/HKAFOs, or wheelchairs (\( M = 2.12 \) and 2.14, respectively), \( F(2, 64) = 4.91, p < .01 \).

**Shunt Surgeries.** For participants who had shunts, Pearson \( r \) correlations were computed to examine relations between number of shunt surgeries...
and the proximal outcomes. Results indicated that number of shunt surgeries was negatively associated with parent-reported scholastic competence, $r = -0.30, p < .05$. These results suggest that children with higher numbers of shunt surgeries exhibited lower levels of scholastic competence. Also, both parent-reported and teacher-reported somatic complaints were positively associated with number of shunt surgeries: parent-report, $r = .46, p < .05$; and teacher-report, $r = .29, p < .05$.

**Composite Severity Variables.** Both the maternal/medical chart composite severity variable and the health professional severity composite were negatively associated with number of parent-reported activities, $r = -0.35, p < .01$, for both. The maternal/medical chart composite severity variable was also negatively associated with both parent- and teacher-reported scholastic competence, $r = -0.28, p < .05$; and $r = -0.34, p < .01$, respectively. Moreover, the maternal/medical chart composite was positively associated with teacher-reported attention problems, $r = .32, p < .01$. Finally, the health professional severity composite was negatively associated with parent-reported athletic competence, $r = -0.26, p < .05$.

**Model B: Severity → Distal Adjustment Outcomes**

Consistent with the hypotheses, the distal adjustment outcomes were not significantly associated with the severity parameters. That is, no significant MANOVA relationships were found between the four categorical condition parameters and the two distal adjustment outcome categories for either parent- or teacher-reported outcomes. However, Pearson $r$ correlation analyses revealed a significant negative association between the maternal/medical chart severity composite variable and the parent-reported Withdrawn subscale of the CBCL, $r = -0.25, p < .05$. The health professional severity composite was also negatively associated with parent-reported social competence, $r = -0.24, p < .05$.

**Models C and D: Indirect and Mediated Effects Models**

Potentially significant model C and D pathways were identified based on the significant univariate model A analyses (which represent the severity→proximal link in models C and D; see results in Table I) and analyses of zero-order associations between the proximal and distal outcomes (which represent the proximal→distal link in models C and D). As would be expected from the model B analyses, the severity→distal direct effects were all nonsignificant; thus, model D was not supported (i.e., mediation was not possible). Thus, the remaining analyses in this section test the significance of model C.

Regarding the zero-order proximal→distal associations, significant associations between these outcomes could be due to common method (respondent) variance because parent and teacher reports are represented in both types of outcomes. To avoid inflating the magnitude of these associations, parent-reported proximal outcomes were examined only in relation to teacher-reported distal outcomes, and teacher-reported proximal outcomes were examined only in relation to parent-reported distal outcomes.

Based on the severity→proximal analyses (see Table I), three proximal parent outcomes (i.e., scholastic competence, activities, and athletic competence) and three proximal teacher outcomes (i.e., attention problems, scholastic competence, and athletic competence) were identified as having significant links with at least one severity variable. Next, zero-order correlations between the three significant proximal parent variables and the seven distal teacher outcomes were examined as were associations between the three significant proximal teacher outcomes and the eight distal parent outcomes. Based on the significant univariate findings in the model A analyses and the significant zero-order proximal→distal effects, pathways were identified where both zero-order links were significant. For these pathways, total effects among the variables were decomposed (via multiple regression) into direct and indirect effects using path analytic procedures (Cohen & Cohen, 1983). Three model C pathways (see Figure 3) met the following four path-analytic criteria: (1) the zero-order severity→proximal and proximal→distal effects were significant, (2) the zero-order proximal→distal effect remained significant after accounting for the contribution of the severity variable (a requirement when computing proximal→distal direct effects with path analysis), (3) the severity→distal direct effect was nonsignificant, and (4) the severity→distal indirect effect was larger than the severity→distal direct effect.
Discussion

Our study investigated relations between condition severity and psychosocial functioning by (1) expanding the set of condition variables examined in past research, (2) disentangling proximal functional status outcomes from distal adjustment outcomes, and (3) using multiple informants for the proximal and distal outcomes. Direct, indirect, and mediated effects models were tested (see models A to D in Figure 2). Findings revealed that the condition parameters were significantly associated with the proximal functional status variables (i.e., Model A). In fact, 9 of the 16 proximal outcome MANOVAs were significant. Moreover, because significant results were found for both parent and teacher report of outcome, it appears that the observed effects generalize across home and school settings. Finally, the results supported an indirect effects model of associations between condition severity and adjustment (Model C) insofar as shunt status was associated with lack of scholastic competence and more attention problems, which were, in turn, associated with social problems and aggressive behavior (see Figure 3).

The lack of findings between the severity variables and the distal adjustment outcomes is consistent with results of past research (Thompson, Gustafson, Hamlett, & Spock, 1992; Varni, Rubenfield, Talbot, & Setoguchi, 1989; Wallander, Feldman, et al., 1989), particularly for internalizing and externalizing symptoms. Although we also failed to find significant associations between severity and social adjustment, findings for this outcome have been mixed in past research (e.g., Wallander, Varni, et al., 1989; see Thompson & Gustafson, 1996, for a review). Significant effects may be found in some studies because certain aspects of social functioning (e.g., the ability to have frequent contacts with friends) may be directly hindered by level of condition severity in some pediatric populations. In fact, some have argued that social adaptation is one component of a child’s functional status (Dadds et al., 1995). We recommend that the social context receive more attention in future severity research.

Consistent with the predictions of this study, findings based on parent- and teacher-report for the physical/athletic proximal outcomes suggest that children with more severe forms of spina bifida (e.g., myelomeningocele with thoracic level lesions) who require more visible forms of aid (e.g., wheelchairs) exhibit lower activity levels and less athletic competence than children with milder forms of spina bifida. Stated differently, and given findings for the distal adjustment outcomes, children with more severe physical disabilities appear to be at-risk for poorer adjustment only in areas directly affected by their physical functioning (also see Billings, Moos, Miller, & Gottlieb, 1987).

Also consistent with the predictions of this study, children with shunts demonstrated more attentional problems and lower levels of scholastic competence than children without shunts, with scholastic competence being even lower in shunted children with higher numbers of shunt surgeries. These results are in line with other reported findings (e.g., Fletcher et al., 1995; Holmbeck & Faier-Routman, 1995; see Wills, 1993, for a review). In the Fletcher et al. study, for example, children with hydrocephalus scored lower on several measures of cognitive functioning. In comparing across studies, however, it is important to recognize that shunted children are not necessarily the only individuals with hydrocephalus; nonshunted children with spina bifida can have compensated hydrocephalus (Holmbeck & Faier-Routman, 1995).

The findings discussed thus far for the proximal outcomes have implications for the noncategorical approach regarding associations between condition severity and psychosocial adjustment (e.g., Stein & Jessop, 1989). From this perspective, adjustment is expected to vary as a function of severity parameters that can be assessed across illnesses (e.g., the degree to which the condition is life-threatening; Thompson & Gustafson, 1996). Although we did not attempt to assess noncategorical severity parameters, our results suggest that there is some benefit to examining condition-specific severity parameters and their relevance for condition-specific functional status outcomes. By adopting an exclusively noncategorical approach, one would fail
to identify associations such as those that emerged in this study. A comparison of the predictive utility of both types of severity parameters across multiple types of pediatric conditions would be a useful direction for future research.

In addition to examining the direct effects of severity on the proximal and distal outcomes, indirect and mediated effects models were also tested (models C and D in Figure 2). The findings were consistent with an indirect effects model rather than the mediated effects model. Such results provide some support for Wallander and Varni’s (1995) contention that severity may be linked with adjustment via the impact that severity has on the functional status of the child. The most robust indirect pathway (after controlling for common respondent variance) involved shunt status, the cognitive proximal outcomes, and the social adjustment distal outcomes. Thus, although shunt status had no significant direct effect on any of the distal outcomes, shunt status was related indirectly with social problems via cognitive functioning. Why was this particular pathway significant? It may be that children with spina bifida who also have problems in school and difficulties with inattention are more likely to be viewed negatively by their peers than are children with spina bifida who are successful in school. Past research supports connections between scholastic competence and social functioning, but investigations of these associations have typically examined this relationship in the direction of social problems to scholastic competence (Cairns, Cairns, & Neckerman, 1989; Elmen, 1991). Although we have specified that the direction of the pathway is shunt→scholastic competence→social problems (see model C), our findings were based on cross-sectional data. Thus, it is not possible to determine the direction of the scholastic competence → social problems portion of the model.

Despite demonstrating support for the proximal and indirect effects models, our study is not without limitations. First, the generalizability of these findings to children and adolescents of other ages is limited due to the narrow age range targeted in the present investigation (8- and 9-year-olds). The age of the sample also may have limited the number of significant results as well. That is, it is possible that the participants had not yet reached the age at which behavioral psychopathology and/or difficulties in social functioning appear (particularly in light of findings that suggest an increase in some internalizing symptoms in adolescence, especially for girls; Nolen-Hoeksema & Girgus, 1994). As these children transition into adolescence, some may be at increased risk for displaying such distal adjustment problems; an increase in variability for these outcomes may lead to more significant associations with condition severity for older children.

Second, 16 families were excluded from participating because they did not speak English (most were Spanish-speaking). Thus, the results of the current study are generalizable primarily to white, English-speaking families. Future studies should include a more representative sampling of Spanish-speaking families to attend to this issue of external validity, particularly given the high rate of spina bifida in Hispanic populations (Lary & Edmonds, 1996). Third, the proximal and distal outcomes were assessed with subscales drawn from the same measures (see Figure 1), which may have inflated associations between these outcomes in the model C analyses. On the other hand, we dealt with some of the common method variance issues in these analyses by only examining associations between proximal parent-reported variables and distal teacher-reported variables or associations between proximal teacher-reported variables and distal parent-reported variables.

Finally, although significant severity→outcome associations were found, very few of the participants demonstrated clinically elevated scores on any of the outcomes (see Table I). Although children who are more severely affected tend to display poorer cognitive and athletic/physical functioning than their less severely affected peers, the former group is still demonstrating subclinical levels of difficulties in these areas. Such moderate elevations in maladjustment are often found in studies of pediatric populations (Wallander & Thompson, 1995).

From a clinical perspective, the findings of this study identify ways in which variations in condition severity manifest themselves directly and indirectly in observable behavioral outcomes. The results suggest that certain pre-adolescents with spina bifida are at risk for adjustment difficulties. Specifically, shunted children with spina bifida are more likely to exhibit significant scholastic and attention problems; children with these problems are, in turn, at particular risk for social adjustment problems. Although children with more severe physical impairments may be less likely to become involved in activities and engage in athletic pursuits, these less active youngsters are not necessarily at risk for other adjustment difficulties. Thus, it appears that
different initial pathways may lead ultimately to different adjustment outcomes. Pediatric psychologists who work with these children will want to be aware of the behavioral consequences which are (and are not) associated with individual differences in the severity of physical disabilities among children with spina bifida.

Acknowledgments

Completion of this article was supported by Social and Behavioral Sciences Research Grants 12-FY93-0621, 12-FY95-0496, and 12-FY97-0270 from the March of Dimes Birth Defects Foundation and a Research Support Grant and paid-leave from Loyola University of Chicago. We thank Ann Walsh Cernak, Caroline Anderson, Joy Ito, Pat McGovern, Pat Braun, David Mc Lone, John Lubicky, the Illinois Spina Bifida Association, and the staff of the spina bifida clinics at Children’s Memorial Hospital, Shriner’s Hospital-Chicago Unit, and the Loyola University of Chicago Medical Center. We also thank Pam Bell, Mariana Belvedere, Rachel Bowers, Michelle Christensen, Ann Marie Czerwinski, Ken Davison, Carrie Donati, Lorin Gorey-Ferguson, Christine Gruse, Jane Holper, Sharon Johnson, Eva Kung, Mai Le, Wendy McKernon, Tina Mikopoulos, Laure Paskiewicz, Anna Rosete, Wendy Shapera, Jennifer Uhler, Venette Westhoven, Christine Wience, and Greer Zummo for help with data collection and data entry. Most important, we thank the parents and children who participated in our study.

Received December 12, 1997; accepted November 12, 1998

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